

Salmonella Gastroenteritis Complicated with Bacteremia and Ruptured Cholangitis in an Infant with Congenital Choledochal Cyst

Chi-Yin Lai, Li-Tung Huang, Sheung-Fat Ko,¹ Jiin-Haur Chuang,² Jui-Wei Lin,³ Mao-Meng Tiao*

Choledochal cyst perforation associated with bacteria is very rare. An 11-month-old infant was admitted to hospital because of fever and bloody diarrhea. Abdominal ultrasonogram revealed a choledochal cyst. Despite antibiotic treatment with initial improvement, jaundice and abdominal tenderness developed 6 days later. Ultrasonography-guided aspiration yielded bile-stained ascites. Emergency operation confirmed the diagnosis of choledochal cyst with perforation. Blood, stool, and bile juice cultures all yielded *Salmonella typhimurium*. To our knowledge, it is rare that a child with choledochal cyst has systemic infection with *S. typhimurium* and cyst perforation. We propose that systemic *Salmonella* infection carries a risk of cyst perforation in patients with congenital choledochal cyst. [J Formos Med Assoc 2007;106(3 Suppl):S20–S23]

Key Words: choledochal cyst, perforation, *Salmonella*

Choledochal cyst is a congenital anomaly of the dilatation of bile ducts,¹ which may present in an acute manner due to spontaneous rupture.^{1,2} Rupture of choledochal cysts associated with blunt trauma^{3,4} and pregnancy⁵ has been described. Here, we report an unusual case of ruptured choledochal cyst complicated with *Salmonella typhimurium* bacteremia. Despite initial clinical improvement in response to antibiotic therapy, acute abdomen culminated in the necessity for emergency operation. To our knowledge, such unusual presentation of choledochal cyst perforation following systemic *Salmonella* infection has not been previously documented.

Case Report

An 11-month-old infant was admitted to the hospital because of bloody diarrhea and fever for 2

days. She was not irritable or crying, and did not have vomiting or consciousness change. Physical examination revealed a weak and febrile baby with a body temperature of 39°C, pulse of 135 beats/minute, respiratory rate of 40 breaths/minute, and blood pressure of 80/52 mmHg. A severe distended abdomen and hypoactive bowel sound were noted upon examination.

Laboratory examinations showed leukopenia (white blood cell count, 1800/mm³), anemia (hemoglobin, 8.8 g/dL; normal, 11–16 g/dL), prolonged prothrombin time (22.4 seconds; normal, 10–14 seconds), elevated C-reactive protein (127.3 mg/L; normal, < 5 mg/L), decreased serum albumin (1.8 mg/dL; normal, 3.0–5.0 mg/dL), elevated aspartate aminotransferase (94 U/L; normal, 13–40 U/L), and alanine aminotransferase (147 U/L; normal, 7–40 U/L). Serum total bilirubin was 1.2 mg/dL (normal, 0.2–1.4 mg/dL), and direct bilirubin was 0.3 mg/dL (normal,

©2007 Elsevier & Formosan Medical Association

Departments of Pediatrics, ¹Radiology, ²Pediatric Surgery, and ³Pathology, Chang Gung Memorial Hospital–Kaohsiung Medical Center, Chang Gung University College of Medicine, Kaohsiung, Taiwan.

Received: May 9, 2006

Revised: June 14, 2006

Accepted: August 1, 2006

*Correspondence to: Dr Mao-Meng Tiao, Department of Pediatrics, Chang Gung Memorial Hospital, 123 Ta-Pei Road, Niasung, Kaohsiung, Taiwan.

E-mail: pc006581@yahoo.com.tw



Figure 1. (A) Abdominal ultrasonography on day 1 of hospitalization shows a dilated common bile duct with a diameter measuring 1.42 cm. (B) Follow-up ultrasonography shows an increased amount of ascites (large arrow) and choledochal cyst (small arrow). (C) Abdominal computed tomography shows a type I choledochal cyst with focal obliteration of the wall (large arrow) suggesting focal rupture with adjacent fluid collection (arrowhead) and minimal ascites adjacent to the spleen (small arrow).

0–0.4 mg/dL). Plain supine and left decubitus abdominal radiographs showed dilated bowel gas but there was no free air. Abdominal ultrasonography showed a dilated common bile duct with diameter measuring 1.42 cm (Figure 1A). The results of clinical and radiologic examination strongly indicated the presence of a type I choledochal cyst. Intravenous cefotaxime (100 mg/kg/day) was administered, with fever and bloody diarrhea subsiding on day 2 of hospitalization. Abdominal distension regressed gradually. Blood and stool cultures all yielded *S. typhimurium*, which was determined to be sensitive to cefotaxime. However, icterus (serum total bilirubin, 4.1 mg/dL; direct bilirubin, 1.6 mg/dL) and abdominal tenderness developed on day 6 of hospitalization. Follow-up ultrasonography and abdominal computed tomography showed an increased amount of ascites (Figure 1B and C). Ultrasonography-guided paracentesis was performed, which yielded bile-stained peritoneal fluid. Analysis of the bile-like peritoneal fluid showed albumin

2.0 mg/dL, amylase 16 U/L, lipase 85,140 U/L, total bilirubin 16.7 mg/dL, and direct bilirubin 9.7 mg/dL. Perforation of choledochal cyst and peritonitis were impressed. An emergency operation was performed and 1000 mL of bile-stained fluid was drained. A small perforation was found on the posterior wall of the choledochal cyst close to the common cystic duct (Figure 2). The culture of bile from the choledochal cyst also revealed *S. typhimurium*. Intraoperative cholangiography confirmed the diagnosis of type I choledochal cyst. The choledochal cyst was excised and a Roux-en-Y hepaticojejunostomy was constructed. The patient was discharged on day 7 postoperatively. She recovered uneventfully and appeared healthy over 12 months of follow-up.

Discussion

Choledochal cyst is a congenital anomaly with focal cystic dilatation of the common bile duct,

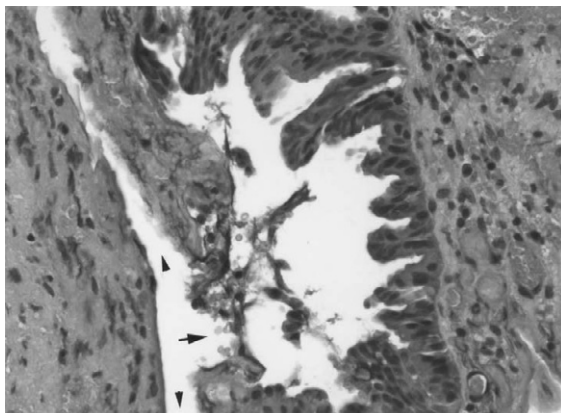


Figure 2. Histologic section of the choledochal cyst shows granulation tissue wall with laceration (arrowheads) and columnar epithelial lining with focal perforation (arrow) (hematoxylin & eosin, 200×).

and is sometimes associated with proximal intrahepatic biliary dilatation. Its etiology remains controversial.^{1–5} Spontaneous perforation was a common complication of choledochal cyst, which was first described in 1934.^{1,2} Different theories of pathogenesis have been proposed, which include pancreaticobiliary system malformation resulting in chronic pancreatic juice reflux into the biliary tract, mucosal abnormality, weakness of the biliary tree walls, or obstruction of the distal common bile duct, etc.^{1,4} To our knowledge, this is the first reported case of perforation of choledochal cyst to be associated with systemic *S. typhimurium* infection.

Acute suppurative cholangitis is a major cause of morbidity and mortality in patients with biliary diseases.⁶ Bacteria, usually from the gastrointestinal tract through the lymphatic system or blood vessels, ascend along the bile duct and cause inflammation of the bile duct.^{6,7} The most common micro-organisms associated with cholangitis were *Escherichia coli* (17.5%), *Klebsiella* spp. (15.7%), *Pseudomonas* spp. (14.6%), and *Salmonella* spp. (5.8%).⁶ Among them, *S. typhi* (53.8%), *S. enteritidis* (17.9%), and *S. typhimurium* (10.7%) were common.⁶ In a previous report, Beiler et al indicated the presence of gallbladder perforations associated with *Salmonella* infection.⁷ In this patient, *S. typhimurium* was isolated from both blood and stool. We believe that this patient might have had enterocolitis at the beginning

but secondary bacteremia and cholangitis supervened. Finally, after swelling and dilatation, the inflammatory choledochal cyst ruptured, causing bile-stained peritonitis. Usually, intravenous antibiotic alone is effective for most *Salmonella* bacteremia;^{8,9} however, emergent surgical intervention should be considered in patients not responding well to antibiotic therapy.^{2,6} In this patient, there was an initial regression in the symptoms of fever, bloody diarrhea, and abdominal distension, presumably due to the *Salmonella* infection responding to antibiotic treatment. This improvement in a patient's clinical status often precludes the capability of the physician to make a diagnosis of the emergent choledochal cyst perforation. In light of this case, we suggest that in patients with *S. typhimurium*-infected choledochal cyst, perforation may be imminent, even with initial clinical improvement following therapy with antibiotics.

Surgery is the treatment of choice for choledochal cyst.^{2,10} Roux-en-Y hepaticojejunostomy, as performed in our case, has been reported to be the technique of choice for biliary reconstruction during the surgical repair of choledochal cyst.^{2,10}

In conclusion, we have reported an unusual case of choledochal cyst associated with *S. typhimurium*, which culminated in eventual rupture of the choledochal cyst. Cautious antibiotic treatment of infected choledochal cyst is important, and if possible, early surgical repair is recommended.

References

1. Ando H, Ito T, Watanabe Y, et al. Spontaneous perforation of choledochal cyst. *J Am Coll Surg* 1995;181:125–8.
2. Ando K, Miyano T, Kohno S, et al. Spontaneous perforation of choledochal cyst: a study of 13 cases. *Eur J Pediatr Surg* 1998;8:23–5.
3. Raj JP, Walsh M. Choledochal cyst and blunt trauma—case report. *Injury* 2002;33:644–6.
4. Ellis H, Cronin K. Bile peritonitis. *Br J Surg* 1960;48:166–9.
5. Hewitt PM, Krige JE, Bornman PC, et al. Choledochal cyst in pregnancy: a therapeutic dilemma. *J Am Coll Surg* 1995;181:237–40.
6. Vaishnavi C, Singh S, Kochhar R, et al. Prevalence of *Salmonella enterica* serovar Typhi in bile and stool of patients

with biliary diseases and those requiring biliary drainage for other purposes. *Jpn J Infect Dis* 2005;58:363–5.

7. Beiler HA, Kuntz C, Eckstein TM, et al. Cholecystolithiasis and infection of the biliary tract with *Salmonella* Virchow—a very rare case in early childhood. *Eur J Pediatr Surg* 1995;5:369–71.
8. Gradel KO, Schonheyder HC, Pedersen L, et al. Incidence and prognosis of nontyphoid *Salmonella* bacteraemia in Denmark: a 10-year county-based follow-up study. *Eur J Clin Microbiol Infect Dis* 2006;25:151–8.
9. Wang JY, Hwang JJ, Hsu CN, et al. Bacteraemia due to ciprofloxacin-resistant *Salmonella enterica* serotype choleraesuis in adult patients at a university hospital in Taiwan, 1996–2004. *Epidemiol Infect* 2006;29:1–8.
10. Bismuth H, Krissat J. Choledochal cystic malignancies. *Ann Oncol* 1999;10:94–8.